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Letter to the Editor

Further Study of the Therapy for Fibrous Dysplasia Is Necessary

To the Editor:

While Dr. Weinstein's clinical experience with bisphosphonate therapy of fibrous dysplasia has been favorable, the evidence presently available cannot justify the conclusion that this "previously medically untreatable bone disease is now amenable to aminobisphosphonate therapy."[1] Although we share his optimism, we feel the issues of medical therapy for this devastating disease deserve more space than may have been available in Dr. Weinstein's article.

To place this study in context, Dr. Weinstein's patient has responded better than many early patients given calcitonin, mithramycin, and the bisphosphonates clodronate and etidronate.[2–5] Despite modest biochemical evidence of effect, none of these case reports described pain control, stabilization/improvement of bone structure, or fracture prevention.

Only with the more potent bisphosphonates has evidence of effectiveness been seen. Liens et al. used pamidronate in 9 patients.[6] Consistent with Dr. Weinstein's experience, bone turnover decreased, and pain was lessened in 12/14 lesion sites. However, radiological improvements were only reported in 4/9 patients, and one child developed a rachitic mineralization defect of the growth plate. In a similar pamidronate trial, Bone et al. confirmed a variable improvement in pain and stabilization of lesion expansion with some evidence of lytic repair and occasional progression.[7]

Despite this trend of evidence, the fact that none of the above studies have been blinded or controlled begs attention. The natural history of fibrous dysplasia in any individual may be subclinical, waxing and waning, or progressive.[8] Thus a properly blinded, randomized, and controlled study will be required to prove effectiveness. Until these data are available, the uncertainty of benefit and the risks of such therapy, particularly in growing children, should be fully described to patients and their parents.

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